The Odontogenic Keratocyst: clinical, radiological, and histopathological study

الكيس المتقرن في عظم الفك: دراسة سريرية، اشعاعية وسريرية

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الخلاصة:

الكيس المقرن في عظم الفك هو مرض حميد ينشأ من الخلايا الجرثومية السنية في عظم الفك. طبقت هذه الدراسة على هذا النوع من الاكياس الفكية سريريا، اشعاعيا ونسيجيا في أرشيف مختبر أمراض الفم / كلية طب الاسنان / جامعة بغداد على اثنين وخمسين مريضا مصابا بهذا الكيس خلال السنوات ١٩٩٠-٢٠٠٠م. أظهرت النتائج ان معدل اعمار المرضى هو (٢٩.٢) سنة، يصاب به الذكور اكثر من الاناث وبمعدل ١:١.٦ . اكثر الحالات سجلت في الفك السفلي (منطقة الاضراس) تليها الضواحك. كان انتفاخ عظم الفك هو ابرز علامة سريرية سجلت في هذه الدراسة. اشعاعيا، ظهر المرض على شكل عتمة شعاعية متعددة المواقع وبنسبة (٦٣.٦%)، رافقها اسنان مطمورة بنسبة (٣١.٨%). نسيجيا، تبين انه في ثلاث حالات فقط تغير النسيج الطلائي المبطن للكيس الى خلايا سرطانية حميدة.

Abstract:

The odontogenic keratocyst of the jaw refers to an uncommon benign lesion that originates from dental primordial. A retrospective study was conducted about the odontogenic keratocyst clinically, radiographically, and histopathologically in a fifty-two Iraqi patients from the archives of Oral pathology department, College of Dentistry, University of Baghdad, over a period from 1990-2000. The result showed that the mean age was (29.2) years, males were affected more than the females with a ratio (1.6:1). The mandibular molar region was the most specific site involved, followed by mandibular premolar area. The most common clinical symptom was the alveolar bone expansion. Radiographic feature revealed a dominancy of multilocular appearance in (63.6%), associated with unerupted tooth in (31.8%). Histologically, the cyst shows ameloblastic variation in only three cases.

Introduction:

The odontogenic keratocyst of the jaws refers to an uncommon benign lesion that originates from dental primordial. It is of particular interest because its aggressive cyst known show rapid growth and its tendency to invade the adjacent tissues, including bone. It has a high recurrence rate. The odontogenic

keratocyst was first described as a distinct entity by Shear in 1960. Philipson originally applied the name “odontogenic keratocyst” to this lesion [1].

The odontogenic keratocyst is classified as a developmental odontogenic epithelial cyst and comprise approximately 8-11% of all cysts of the jaws [2, 3].

In study conducted on 255 patients, Ahlfors et al. [4] found an average age at the time of diagnosis of 41 years; while Chow [5] reports that the majority of the patients were 21 to 30 years of age. They were more common in men particularly in the second and third decades, with (72.2%) of the cases were in men [6].

The mandible is the most frequent site for the odontogenic keratocyst, about 50% occurs at or near the angle although they may be present anywhere in the jaw [7]. The molar region of either the mandible or maxilla was the principle primary location, the maxillary antrum was also a common site [8].

Clinically, an odontogenic keratocyst is characterized by aggressive, local growth. The lesion may manifest with pain, swelling, discharge, and occasionally parasthesia or displacement of teeth [4]. The clinical significance of the odontogenic keratocyst is the potential for morbidity associated extensive expansion and recurrence after surgery [9].

Radiographically, the odontogenic keratocyst is typically well demarcated and unilocular or multilocular lesion, teeth and bony structure such as the inferior alveolar canal may be displaced, root resorption has been reported; the prevalence of which has varied from rare to affecting 24% of all teeth associated with odontogenic keratocyst [10, 11]. Associated with an impacted mandibular molar in more than 50% of the cases [5].

The histopathological characteristic of the odontogenic keratocyst have been well defined and are widely recognized its epithelial lining generally exhibits a uniform thickness and is usually parakeratinized although an orthokeratinized type also occur. It exhibits a distinctly palisade basal layer of hyperchromatic cell. The connective tissue wall often shows small islands of epithelium similar to the lining epithelium; some of these islands may be small cyst “daughter cyst” [12].

The purpose of this study is to find the incidence, clinical features, radiographical appearance and histopathological variation of odontogenic keratocyst in a series of odontogenic keratocyst of 10 years. Such information is valuable to clinician as it helps in the formulation of a working diagnosis and timing management decisions and approach to treatment.

Material and Methods:

All cases of odontogenic keratocyst over the period between 1990-2000 were recorded from the laboratory of Oral pathology, College of Dentistry, University of Baghdad.

The clinical, radiologic, and pathological details were taken from the archives file of the Oral pathology department. All cases diagnosed histologically as odontogenic keratocyst were analyzed according to the age, sex, duration, site distribution, clinical presentation, radiographic appearance and histopathologic details based on the individual pathologic report of each case. These files more checked for adequacy of information given by the surgeon regarding clinical presentation, site of cysts and other information regarding the case at question. This information's were tabulated.

Results:

From the total of 2410 oral biopsies, 235 cases were diagnosed as jaw bone cysts, fifty-two cases were diagnosed as odontogenic keratocyst, accounting (22.1%) of all jaw bone cysts, and (2.2%) over all oral surgical biopsies.

The age of the patients ranged from 10 to 7o years, with a peak incidence in the third decade of life (19) cases (36.6%), and the mean age was (29.2%) years. The males were affected more than the females with a ratio of (1.6:1). However, the maximum male predominance was observed in the sixth decade (4:1). Table 1.

Table 1: Age and sex distribution of 52 cases of odontogenic keratocyst

Age group (years)	Number	Male	Female	Male:Female ratio
1-10	1(1.9%)	1(1.9%)	0	1:0
11-20	6(11.6)	3(5.8%)	3(5.8%)	1:1
21-30	19(36.6%)	12(23.2%)	7(13.6%)	1.7:1
31-40	11(21.1%)	6(11.6)	5(9.6%)	1.2:1
41-50	6(11.6)	4(7.6%)	2(3.8%)	2:1
51-60	5(9.6%)	4(7.6%)	1(1.9%)	4:1
61-70	4(7.6%)	2(3.8%)	2(3.8%)	1:1
Total	52(100%)	32(61.5%)	20(38.5%)	1.6:1

Regarding the site distribution, 36 cases (69.2%) occurred in the mandible and 16 cases (30.8%) in the maxilla, giving the mandibular to maxillary ratio of 2.3:1. In nine cases the specific region was not recorded. However, for the mandible, the most common affected site was molar region (17 cases = 32.7%), followed by premolar region (8 cases = 15.4%). While for the maxilla the most common affected site was also molar region (7 cases = 13.5%), followed by incisor region (4 cases = 7.7%). However, the lesion crossed the midline in four cases (7.7%), and extended to the ramus area in 7 cases (13.5%). Table 2.

Table 2: Site distribution of 52 cases of odontogenic keratocyst.

Jaw	Incisor + Canine	Premolar	Molar	Unknown	Total
Mandible	6 (11.5%)	8(15.4%)	17(32.7%)	5(9.6%)	36(69.2%)
Maxilla	4(7.7%)	1(1.9%)	7(13.5%)	4(7.7%)	16(30.8%)
total	10(19.2%)	9(17.3%)	24(46.2%)	9(17.3%)	52(100%)

The duration of the symptoms ranged from two months to three years (median = 8 months). The initial presenting symptom for all 52 patients was expansion of the jaw bone, associated with pain in fifty cases; tooth displacement was noted in eight cases, while parasthesia was observed in four cases. Furthermore, only two cases were recorded as recurrent cases from the reports of the patients.

The radiologic appearances of odontogenic keratocyst are demonstrated in table 3. Excluding of eight cases (15.4%) because the radiographic details were not available. In 28 cases (63.6%) the lesion had a multilocular radiolucent appearance, in 16 cases (36.4%) it was unilocular radiolucency. However, in the mandible 19 cases (43.1%) of the lesions were multilocular in compared with 9 cases (20.5%) in the maxilla. In addition the odontogenic keratocyst was associated with an unerupted tooth in 14 cases (31.8%), causes root resorption in 12 cases (27.3%), and involved the maxillary antrum in only one case (2.8%).

Table 3: Radiographic appearance of 44 cases of odontogenic keratocyst

Jaw	Multilocular	Unilocular	Impacted tooth	Root resorption
Mandible	19(43.1%)	12(27.3%)	11(25%)	7(15.9%)
Maxilla	9(20.5%)	4(9.1%)	3(6.8%)	5(11.4%)
Total	28(63.6%)	16(36.4%)	14(31.8%)	12(27.3%)

All of the pathologic reports describe a similar histologic picture. The lesion consist of a fibrous connective tissue wall which contain islands of epithelium shows a small cystic lesion “daughter cyst”. The epithelial lining is highly characteristic, and is composed of (1) a parakeratin surface which is usually corrugated or wrinkled, (2) a uniformity of thickness, generally between 4 to 10 cells in depth without rete peg, and (3) a palisaded basal layer of cells. Occasionally, orthokeratin is observed in some cases. The lumen of the cyst may be filled a thin straw-colored fluid or with a thicker creamy material. Sometimes, the lumen contains a great deal of keratin. Cholesterol and hyaline bodies at the site of inflammation may also be present. Finally, dysplastic and neoplastic transformation of the lining epithelium were not recorded except in three cases in which the ameloblastic changes observed within the lining epithelium.

Discussion:

The developmental odontogenic cysts occur rarely in the jaw bones as compared to inflammatory cysts. However, the present study revealed that the odontogenic keratocyst is one of the most common jaw bone cyst (22.1%), this finding is higher than that conducted by Shear [(2)] and Moody et al. [(3)].

They are reported to be occurring most often in the third decade of life [(5)]. This is in harmony with our series in which the half of our patients was young, 26 out of 52 patients being under the age of 30 years (50%). However, Neville et al. [(13)] reported in study conducted on 18 cases of odontogenic keratocyst a mean age of 69.9 years, which is much higher than for odontogenic keratocyst.

Concerning the sex distribution, the present investigation showed that males were affected more than females. A similar male predominance also reported by

Nohl and Gulabivala [6], and Neville et al. [13]. This finding is in contrast with that reported by Chung et al. [1].

Regarding the site distribution, odontogenic keratocyst in our study occurs predominantly in the mandibular molar area. These finding confirmed by previous studies [7, 8]. However, Neville et al. [13] reports 18 cases of odontogenic keratocyst occur in anterior midline of the maxilla, and suggest that it is important to include odontogenic keratocyst in the differential diagnosis of anterior midline maxillary radiolucency, especially when they occur in older individuals. Moreover, in our study, four cases of odontogenic keratocyst crossed the midline, five cases extend to the ramus region, and only in one case the lesion involved the maxillary antrum. High incidence of maxillary antrum involvement was reported by Meara et al. [8]. However, Chehade et al. [14] reported six cases of peripheral odontogenic keratocyst, and a case of intracranial invasive odontogenic keratocyst observed by Franc et al. [15].

The odontogenic keratocyst is typically well demarcated unilocular or multilocular lesion, associated with an impacted tooth in more than 50% of the cases [5, 10, 11]. This finding is supported by our series, in which the cysts show multilocularity in 63.6%, and unilocularity in 36.4%. However, the prevalence of association with an unerupted tooth were 31.8%, and root resorption in 27.3%. Moreover, Struthers and Shear [10] reported that the root resorption by odontogenic keratocyst appears to be very rare in comparison with that associated with dentigerous cyst.

Clinically, odontogenic keratocyst occur most often as painless alveolar swelling[4],sometime associated with pain [9] . Our finding is the same, in which the majority of the cases were seen in advanced stages, pain reported in 15 cases (29.6%), while tooth displacement and parasthesia were observed in some cases. The clinical significance of the odontogenic keratocyst is potential for morbidity association with extensive expansion and recurrence rate after surgery [9, 15]. However, in our study only two cases were recorded as a recurrent case.

The histopathological features of our odontogenic keratocysts are similar with those seen elsewhere [12, 17]. Very rarely squamous cell carcinoma has been found arising in odontogenic keratocyst [6]. However, a case of odontogenic keratocyst with chondroid in the cyst wall was reported by mosqueda et al. [16]. A diagnosis based solely on clinical information can be problematic. In many cases, histologic examination of surgically removed tissue is necessary to establish a

definitive diagnosis [17]. In this study only three cases show ameloblastic characteristic within the lining of odontogenic keratocyst.

However, the odontogenic keratocyst (OKC), now officially known as the keratocystic odontogenic tumour (KCOT); WHO"s reclassification of the OKC as the KCOT based on behaviour, histology and genetics underscores the aggressive nature of the lesion and should motivate clinicians to manage the disease in a correspondingly aggressive manner [18] .

References:

1- Chuong R, Donoff B, Gurlnick W. The odontogenic keratocyst. J Oral Maxillofac Surg 1982; 40:797-802.

2- Shear M. Developmental odontogenic cyst: an update. J Oral Pathol Med 1994; 23:1-11.

3- Moody JB, Quigly N, Sutley SH, Ferguson HW. Odontogenic Keratocyst. Gen-Dent 1997; 45(2):172-6 (Medline).

4- Ahlfors E, Larsson A, Sjogren S. The Odontogenic Keratocyst: A benign cystic tumor? J Oral Maxillofac Surg 1984; 42:10-19.

5- Chow HT. Odontogenic Keratocyst: a clinical experience in Singapore. Oral surg Oral Med Oral Pathol 1998; 86:573-7.

6- Nohl FSA, Gulabivala K. Odontogenic Keratocyst as periradicular radiolucency in the anterior mandible. Oral surg Oral Med Oral Pathol 1996; 81: 103-9.

7- Eliasson S, Isacsson G, Kondell PA. Lateral periodontal cysts: clinical, radiographical, and histopathological findings. Int J Oral Maxillofac Surg 1989; 18:191-3.

8- Meara JG, Shah S, Li KK, Gunningham MJ. The odontogenic Keratocyst: a 20-years clinicopathologic review. Laryngoscope 1998; 2:280-3 (Medline).

9- Rud J, Pindborg JJ. Odontogenic Keratocyst: a follow-up study of 21 cases. J Oral Surg 1969; 27:323-30.

10- Struthers P, Shear M. Root resorption by ameloblastoma and cysts of the jaws. Int J Oral Surg 1976; 5:128-32.

11- Partridge M, Towers JF. The primordial cyst (odontogenic keratocyst): its tumor-like characteristic and behavior. Br J Oral Maxillofac Surg 1987; 25:271-9.

12- Shafer WG, Hine MK, Levy BM. A textbook of oral pathology, 4th ed, Philadelphia: WB Saunders Co, 1983:269-73.

13- Neville BW, Damm DD, Brock T. Odontogenic Keratocyst of the midline maxillary region. J Oral Maxillofac Surg 1997; 55:340-4 (Midline).

14- Chehade A, Daley TD, Wysoki GP, Miller AS. Peripheral odontogenic keratocyst. Oral Surg Oral Med Oral Pathol 1994; 77:494-497.

15- Franc C, Cresseaux P, Richard L, Breton P, Freidal M. The Keratocyst or epidermoid cyst: the current state of understanding apropos of a case with intracranial involvement. Rev-Stomatol-chir-Maxillofac 1996; 97:270-82 (Midline).

16- Mosqueda TA, De-la JM, Trencozo VF. Odontogenic Keratocyst with choodroid fibrous wall. A case report. Int J Oral Maxillofac Surg 1998;27:58-60 (Midline).

17- Mohammad A, Ronald AB. Maxillary odontogenic cyst. A common and serious clinical misdiagnosis. J Am Dent Assoc 2003;134(7):877-883.

18- Madras J, Lapointe H. Keratocystic odontogenic tumor: reclassification of the odontogenic keratocyst from cyst to tumor, J Can Dent Assoc 2008;125(5):446-54.